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Original Communication

An audit of the use of definitions of sudden infant death syndrome (SIDS)

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Abstract

Given that there are a number of contradictions in the SIDS literature and that the definition of SIDS that was relied upon to authenticate cases in reports is not always specified, an audit of publications was undertaken. Fifty papers dealing with SIDS that were published in 2005 were reviewed. The majority (58%) of reports had either not specified a definition of SIDS, or had used non-standard or idiosyncratic definitions. Of the papers that had documented a definition: 30% used the 1989 NICHD definition, 10% used the 2004 San Diego definition, and 2% used the 1969 Seattle definition. Failure to use standard published definitions of SIDS and/or to clearly specify the definition that has been followed may severely hamper the evaluation of SIDS research.

Keywords: Sudden infant death syndrome; Definition; Research; Audit

1. Introduction

Sudden infant death syndrome (SIDS) is still the most common cause of unexpected death in Western communities of infants who were previously thought to be healthy. Death rates have fallen sharply over the previous 15 years following the identification and avoidance of environmental risk factors that were found to significantly increase the risk of SIDS. These include sleeping face down, exposure to cigarette smoke and covering of the head with bedding.[1–3]

A continual problem has been the lack of pathognomonic pathological features in SIDS that has left it a diagnosis of exclusion.[4] In addition, a number of definitions have been promulgated, each of which has emphasized slightly different features, often resulting in considerable confusion as to what constitute the defining characteristics in the minds of researchers and SIDS families alike.[5–10]

SIDS is a phenomenon that has also been extensively investigated and written about, however, the authors have noted that there appears to be a lack of consistency in the use of definitions.[11] Unfortunately, variable case selection and the use of different definitions may have an impact on research results, with considerable potential for bias if uniformity in approach does not occur. The following study was, therefore, undertaken to evaluate the consistency with which definitions are used in SIDS research.

2. Materials and methods

The United States National Library of Medicine 'Entrez PubMed' database[12] was searched for all entries listed under "sudden infant death syndrome" and also for all such entries for the year 2005. Of the 216 papers listed for 2005, 50 were selected from the printed abstracts where the veracity of the conclusions depended on accurately defining SIDS. 'Letters to the Editor', 'Brief Reports' and commentaries that did not specifically focus on SIDS were excluded. The papers were reviewed and checked to determine whether one of three common definitions of SIDS[5–7]

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had been either written in the text or referenced. The specific definitions that were searched for included:

(1) *Seattle definition*: In 1969 at the Second International Conference on Causes of Sudden Death in Infants it was proposed that SIDS was 'the sudden death of any infant or young child which is unexpected by history, and in which a thorough post-mortem examination fails to demonstrate an adequate cause of death'.[5]
(2) *NICHD definition*: In 1989 the National Institute of Child Health and Human Development (NICHD) convened an expert panel to re-examine the issue of definition. The panel proposed that SIDS was 'the sudden death of an infant under one year of age, which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history'.[6]
(3) *San Diego definition*: In 2004 the CJ Foundation convened an expert panel to again re-examine the issue of definition. The panel proposed that SIDS was 'the sudden and unexpected death of an infant under 1 year of age, with onset of the lethal episode apparently occurring during sleep, that remains unexplained after a thorough investigation including performance of a complete autopsy, and review of the circumstances of death and the clinical history'.[7]

Papers that did not cite one of the three definitions above were placed in either of two categories:

(4) those that used idiosyncratic, other or mis-cited definitions, and,
(5) those where there was no explanation of how the cases that were being called SIDS had been defined.

3. Results

Search of the Entrez PubMed database revealed 6,458 entries under 'sudden infant death syndrome' up to November 2006, with 216 papers listed for the year 2005. Within the 50 papers selected from 2005 there was a mixture of original research, case or series reports, meta-analyses and reviews. The pattern of citation and referencing of the definitions is listed below:

Seattle definition:	One paper where it was both written in the text and referenced. ($N = 1$; 2%).
NICHD definition:	Eleven papers where it was both written in the text and referenced, one where it was written in the text but not referenced, and three where it was only referenced. ($N = 15$; 30%).
San Diego definition:	Four papers where it was both written in the text and referenced, and one where it was only referenced. ($N = 5$; 10%).
Idiosyncratic/other/mis-cited:	($N = 8$; 16%).
Not cited:	($N = 21$; 42%).

4. Discussion

The results demonstrate that the majority (58%) of a series of 50 papers dealing with SIDS that were taken from the peer-reviewed literature in 2005 had either not specified the definition of SIDS that was being used, or had used a non-standard definition. Idiosyncratic additions/alterations to definitions included the specification of different age ranges, and recommendations for the necessity, or not, of history review and ancillary investigations. Most papers that documented the definition followed the NICHD guidelines (30%), with the majority quoting the definition in the text as well as citing the appropriate reference. Only 10% of papers cited the San Diego definition and 2% the Seattle definition.

The purpose of this study was not to identify particular papers where a definition of SIDS was being used, or not used, as the latter may have been an oversight that occurred during the time of the collating and writing-up of results. Instead, the intention was to draw attention to an unfortunate situation that exists in the SIDS literature where significant difficulties sometimes arise in attempting to determine the validity of a particular piece of research because of a failure to clearly describe how the initial case classification and accession was undertaken.

Considerable contradiction exists in the SIDS literature on a number of issues,[4] and this may have been contributed to by variations in the initial classification of cases as SIDS. For this reason it is vital for writers and researchers who are publishing data on SIDS to clearly specify how cases were investigated and how the diagnosis of SIDS was arrived at. Lack of a uniform approach among different centres means that comparisons of data may be meaningless, or even impossible. It must also be recognised that merely because a definition has been cited in a paper does not mean that all of the cases analysed have been diagnosed according to the cited criteria.

Accepting that a case fulfils standard criteria for the diagnosis of SIDS merely because it has been listed in a data base as 'SIDS', or has had this label used on an autopsy report, may be of minimal usefulness. One advantage of the San Diego approach is that stratification of cases enables researchers to assess the degree of likelihood that a death was actually due to SIDS. The definition provides a gradient of certainty for the diagnosis.[7] Given the

complexities of many issues in SIDS research, the sooner that researchers can agree to adhere to common parameters and definitions, and to clearly specify how these have been used in the Materials and Methods section of their papers, the greater will be their chances of integrating and validating SIDS research.

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